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CASE REPORT

A bulky dermoid cyst of the floor of the mouth

E. Boko^{a,*}, K. Amaglo^b, E. Kpemissi^a

^a Service d'ORL et de chirurgie cervico-faciale, CHU de Lomé, BP 30006 Lomé, Togo

^b Service d'odontostomatologie, CHU de Lomé, BP 30006 Lomé, Togo

KEYWORDS

Cyst;
Dermoid cyst;
Floor of the mouth;
Congenital tumor

Summary

Introduction: Bulky dermoid cysts of the floor of the mouth are very rare and may pose a problem of diagnosis. They also raise problems for the anesthesiologist and surgeon. We report the first case to be described in Togo.

Case report: A 23-year-old man was admitted for a submental submandibular sublingual mass. It was soft, depressable, painless, without adenopathies, raising the tongue against the palate and creating a "second tongue-like" aspect. Resection on intra-oral route removed an intact cyst of 13 cm long axis. Histology diagnosed dermoid cyst.

Discussion: Dermoid cysts of the floor of the mouth present as a submental sublingual mass, which may cause dyspnea and disorders of swallowing, chewing and/or vocal function. Differential diagnosis concerns sublingual, submental and cervical masses. Definitive diagnosis is founded on the histology specimen. Imaging may assist diagnosis. Intubation may be problematic. The resection approach may be intra-oral or cervical.

Conclusion: Dermoid cysts of the floor of the mouth are rare. They may induce functional disorder. An intra-oral approach is preferable when possible.

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Introduction

Dermoid cysts of the floor of the mouth are rare. They represent 1 to 1.6% of all dermoid cysts [1]. About 225 cases have been reported [1,2]. Bulky forms are even rarer and raise diagnostic problems. They also pose problems for the anesthesiologist and surgeon. We present the first case to be reported in Togo and probably the first in the African French-language literature.

Case report

A 23-year-old man was admitted for a submental submandibular sublingual mass, evolving since birth. Associated

signs comprised muffled voice and snoring. The mass was located submentally, extending into two submandibular regions (Fig. 1a and b), and was soft, depressable, painless and free of adhesions and adenopathies. Intra-oral examination found the mass raising the tongue against the palate, creating a "second tongue" aspect (Fig. 2c). Tongue motion was conserved (Fig. 2a and b). The mucosa, sublingual glands and salivary caruncles were normal. A mucous sublingual gland cyst was suggested. Ultrasound and CT could not be performed, for financial reasons. After minimal pre-operative assessment, surgical resection was undertaken. Intubation was difficult, but had to be performed by nasotracheal route. A mucosal incision was made on the ventral side of the tongue, transfixing the frenulum and extending 3 cm on either side. The mucosa was detached along approximately 4 cm, and dissection was guided mainly by finger touch. The anterior sublingual (Fig. 3a), submental inferior cervical (Fig. 3b) and posterior oropharyngeal extensions

* Corresponding author. Tel.: +22 8 90 04 53 25.
E-mail address: bokonorbert@yahoo.fr (E. Boko).

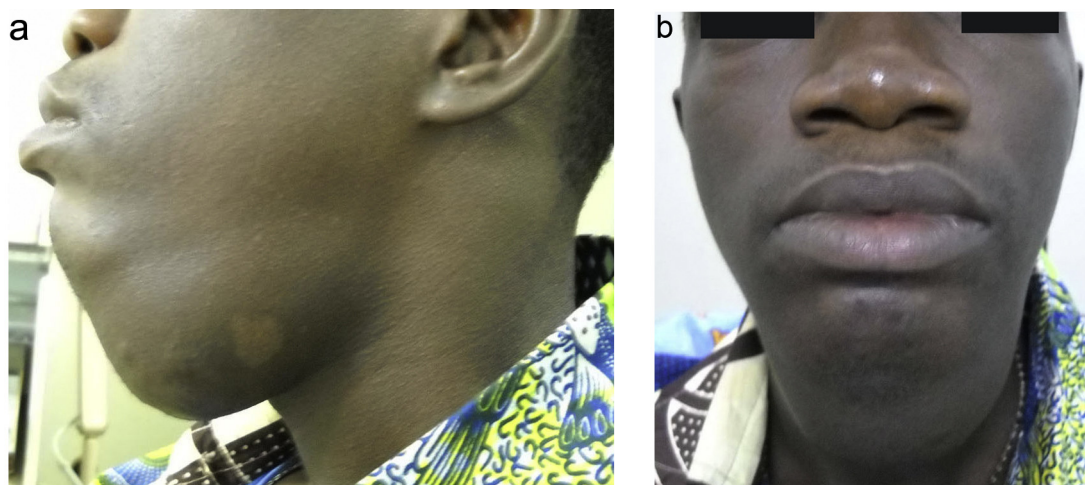


Figure 1 Extra-oral frontal (b) and lateral view (a): bulky submental submandibular mass.

(Fig. 3c) were successively released. A fistulous track toward the left tonsillar fossa (Fig. 3c) suggested an unusual second-arch cyst location. After complete resection, the cyst was removed intact. It measured 13 cm on the long axis. The patient was discharged on day 6. Histology found a dermoid cyst of the floor of the mouth, without malignancy. At 3 months follow-up, there were no sequelae, complications or recurrence.

Discussion

One of the first descriptions of dermoid cyst of the floor of the mouth was in Jourdain's *Traité des Maladies de la Bouche* of 1778 [3]. 6.9% of the 1495 dermoid cysts reported by Erich had head-and-neck locations, and only 1.6% were located on the floor of the mouth [3]. The English-language literature as a whole contains only one case

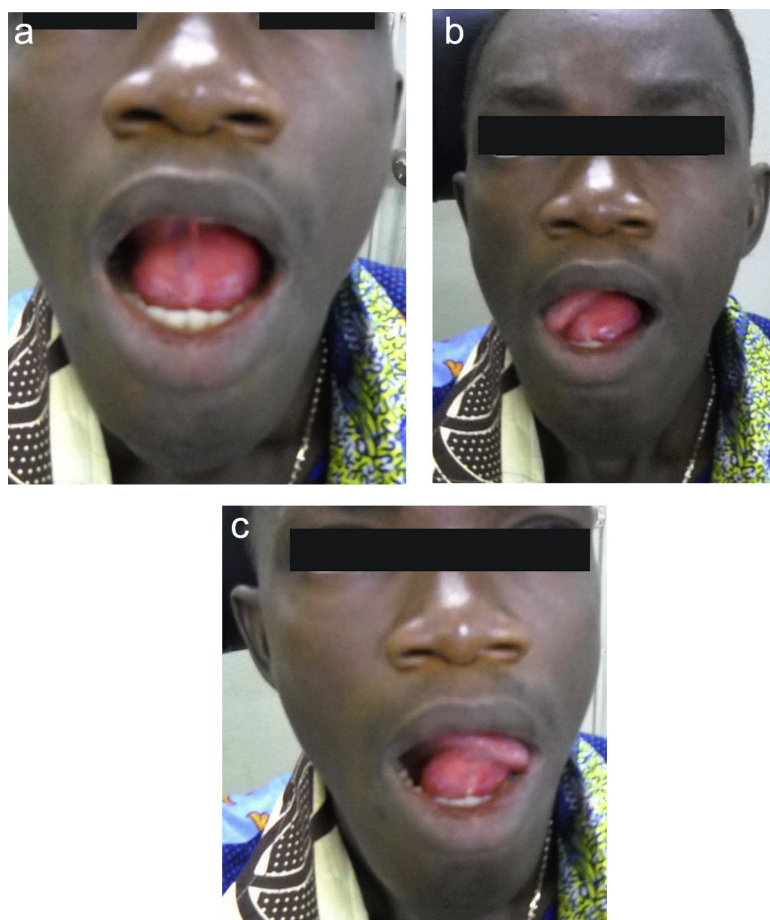


Figure 2 Intra-oral view: tongue motion is conserved (a and b), creating a "second tongue" aspect (c).

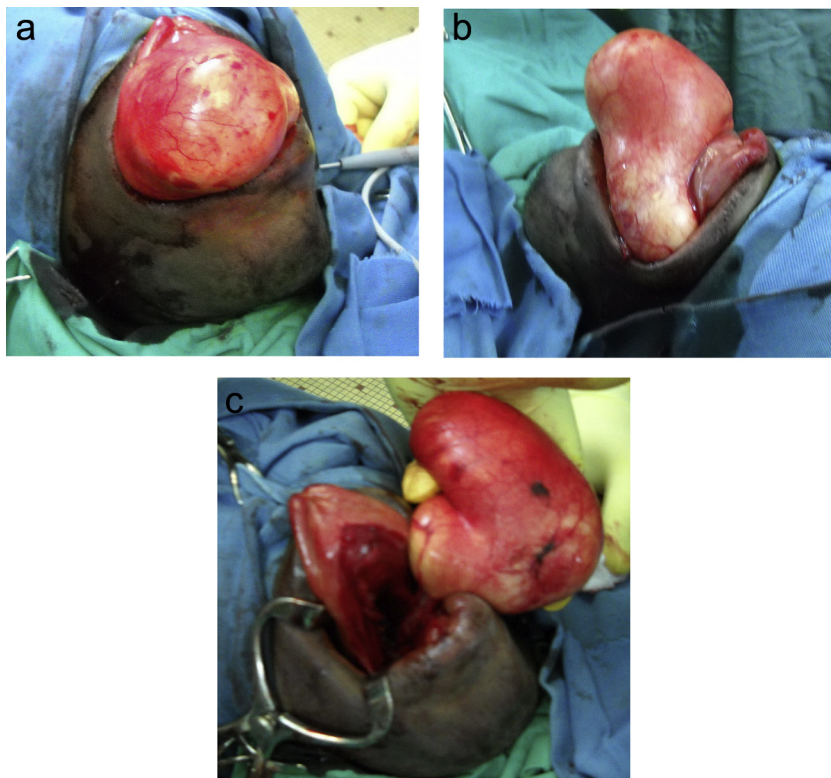


Figure 3 Peroperative view: resection of the anterior (a), inferior (b) and posterior extension with fistulous track (c).

report [4]. The present case is the first to be described in Togo.

Onset is mainly during the second and third decades of life [2] but may be congenital, as is clearly the case in the present patient. Meyer's 1955 classification [5] categorizes dermoid cysts as: epidermoid, true dermoid or teratoid cysts [1].

Dermoid cyst of the floor of the mouth presents as a sublingual submental mass of gradually increasing volume, soft and painless, with possible associated dyspnea and disorders of swallowing, chewing and the voice. Authors have reported sudden increases in volume at puberty, related to increased sebum secretion [6]. Differential diagnosis concerns sublingual, submental and cervical masses. In mucous sublingual gland cyst, aspiration finds mucous content, whereas dermoid cyst content is thick, paste-like, granulated and whitish. Second branchial arch cysts are usually cervical in location: a fistulous track toward the tonsillar fossa seemed at first suggestive of this diagnosis in the present case. Other forms of mass may confuse diagnosis: thyroglossal tract cyst, cystic lymphangioma, ectopic thyroid gland, lipoma, cellulitis, or tumor of the floor of the mouth. Definitive diagnosis is provided by the histologic specimen.

Imaging can assist diagnosis [7]. Ultrasound finds a pseudosolid anechogenic or finely echogenic cystic mass. A fat/liquid line with more echogenic supernatant fluid and/or a more echogenic floating mass are suggestive. CT finds a thin-walled unilocular mass, with fatty content. A low-lying liquid line, moving with change in position, is characteristic. MRI finds a fatty level in hypersignal on T1-weighted

sequences, with fall in signal on FatSat sequences. CT and MRI identify contiguous organs, guiding the choice of surgical approach [8].

Intubation can be difficult when the cyst is very bulky; tracheotomy may be required. Raveenthiran et al. [9] managed a 10 × 10-cm cyst by first emptying it, to facilitate intubation; in our view, however, this could hinder resection, with a risk of leaving some of the cyst wall and thus of recurrence. The surgical approach may be intra-oral or cervical: the former is best suited to small lesions [2], reserving the latter for bulky lesions. Certain authors performed mandibular symphysis osteotomy and a combined intra-oral and cervical approach [2]. An exclusively intra-oral approach (as in the present case) can be used even with bulky lesions if there is no superinfection or functional impairment; it entails a lower risk of postoperative superinfection, shortens the hospital stay, and provides an excellent esthetic result. Recurrence is rare, but may follow incomplete resection. Malignant transformation is exceptional, but has been reported [10]: in 75% of cases it consists of squamous cell carcinoma and is almost systematically invasive [7].

Conclusion

Dermoid cysts of the floor of the mouth are rare, with onset generally at birth. When bulky, they may induce functional disorder. An intra-oral approach is preferable in absence of complications. Postoperative prognosis is good, and malignant transformation is exceptional.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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